Original Article

Norm-referenced scoring system for the Brief Infant Sleep Questionnaire — Revised (BISQ-R)

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ABSTRACT

Objectives: To develop an age-based norm-referenced scoring system for the Brief Infant Sleep Questionnaire — Revised (BISQ-R).
Methods: In sum, 33,835 submissions (data sample 1) of the expanded and revised BISQ-R by caregivers of infants and toddlers (1—36 months) were analyzed in the US via a publicly-available smartphone application, Johnson's® Bedtime®. Three subscales were created: Infant Sleep (IS; 5 items), Parent Perception (PP; 3 items), and Parent Behavior (PB; 11 items). The scoring algorithm was based on an age-based normative system, and each subscale was scored using a weighted average of items. Primary application of the scoring model was performed on a follow up set of 16,531 submissions (data sample 2). Secondary application was tested on an original web-based ecology study (data sample 3). Tertiary application was tested using previously published datasets consisting of a longitudinal study (data sample 4) and randomized control trial behavioral intervention study (data sample 5).
Results: Overall application of the scoring algorithm was confirmed across multiple samples. Each subscale (IS, PP, PB) and total score (T) is age referenced (scores range from 0 to 100). Cross-comparison between subscales across studies reveal consistent and convergent relationships.
Conclusions: The BISQ-R provides a comprehensive assessment of infant and toddler sleep patterns, as well as parent perception and parent behaviors that may contribute to sleep outcomes. The age-based norm-referenced scoring system is publicly available to be used by researchers and clinicians.

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1. Introduction

Sleep is a concern common to parents of infants and toddlers, and 25—30% of parents report their child experiences a sleep problem [1,2]. Many studies has been conducted on parent behaviors that contribute to sleep outcomes in this age group. For example, research has shown that parental presence at bedtime is highly associated with increased sleep disturbances and changes in parental behaviors results in amelioration of infant and toddler sleep disturbances [3—7].

Furthermore, many studies have demonstrated that sleep changes dramatically across the first few years of life [8—10]. In newborns, sleep is characterized by short sleep-wake periods between 1—3 h, with sleep consolidation beginning between three and six months of age. Naps shift from several short naps a day in the first months to typically two longer naps by nine months of age. At approximately 15—18 months of age, most toddlers have shifted to a solo, longer afternoon nap. Thus, unlike in older children, adolescents, and adults, assessments of sleep patterns must be based on very narrow age bands of weeks to a few months.

The Brief Infant Sleep Questionnaire [11,12] (an assessment measure of infant and toddler [0—36 mos] sleep patterns, sleep ecology, and parental perceptions of sleep) has been used in over 50 studies since its first publication by Sadeh in 2004. The original scale has been revised (BISQ-R) to include a wider array of sleep behaviors and outcomes. The questionnaire includes specific items

about infant daytime and nighttime sleep patterns, as well as sleep-related behaviors and parent perception. It has been translated into over 20 languages and has been validated against actigraphy and daily-logs [11]. A recent Spanish adaption of the BISQ-R [13] indicated excellent test-retest reliability, r = 0.848 (P < 0.001), with a kappa value of 0.939 (95% CI: 0.858−1.00, P < 0.001) for agreement between self-administration and clinical interview. Agreement between an earlier expanded version of the BISQ [12] and sleep diary also was demonstrated for bedtime, hours of nighttime and daytime sleep, and number of night wakings (P < 0.001). Furthermore, its sensitivity in documenting expected developmental changes in infant sleep [14] and the effects of environmental factors has been established [15] in previous studies. A drawback of the measure to date, however, has been the lack of a scoring system, which would significantly increase its utility in clinical situations and in research studies.

Thus, the purpose of this manuscript is to describe the preliminary development and application of a norm-referenced age-based scoring system for the BISQ-R. In addition to a total score, three subscales were constructed including Infant Sleep (IS), Parent Perception (PP), and Parent Sleep-related Behaviors (PB). The scoring system was first developed using a large database of 33,835 young children, and then replicated with a second large sample of 16,531 infants and toddlers. Further application of the scoring system was conducted with three additional data sets, one that comprised of a web-based sleep ecology study of 4595 infants, another that followed 164 infants longitudinally, and a third from a randomized control trial of an internet-based behavioral intervention for 264 infants and toddlers with sleep disturbances.

2. Methods

All studies were approved by the Institutional Review Board at Saint Joseph's University. All participants provided consent to the inclusion of their data in research and were able to decline participation.

2.1. BISQ data samples used for development and application of the scoring algorithm

Several data samples were used for the development and application of the scoring algorithm (see Fig. 1). Real world data sample 1 was used for model creation and initial scoring. Real world data sample 2 was used for primary replication. Web-based
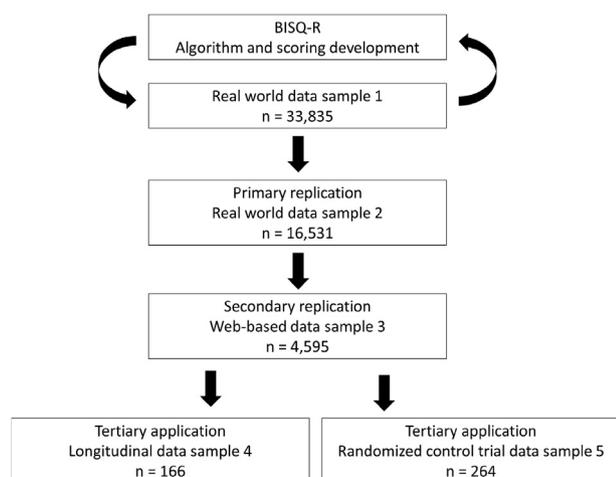

Fig. 1. BISQ-R data samples used for construction, replication, and application of the scoring algorithm.

data sample 3 was used for secondary application. Longitudinal data sample 4, and trial/control intervention data sample 5 was used for tertiary application. Further details about each data sample are described below.

2.2. BISQ-R scoring framework

The BISQ-R includes 33 questions and is completed based on recall of the past two weeks. For scoring, there are three expert-defined, content-based subscales (see Fig. 2). The subscales were conceptually developed to capture three aspects of infant sleep, infant sleep patterns (IS score), parental perceptions of their child's sleep (PP score), and ecologically-based parental behaviors that have been found to impact infant sleep (PB score). These three independent, but interconnected constructs, have been found to be associated with parental behaviors impacting infant sleep (in one study, accounting for 21−39% of the variance) and infant sleep patterns accounting for 22% of the variance in parental perception of sleep problems [12].

2.2.1. Infant sleep subscale

The IS includes five questions regarding sleep patterns, including sleep onset latency, number and duration of night wakings, longest stretch of sleep, and total night sleep.

Item scoring: Scoring of items in this subscale is derived from age-based normative US data (real-world data sample 1). The distributions for each IS item were calculated and mapped to a normative scale from 0 to 1 (0.1 being ~10%, 0.5 being ~50%, 0.9 being ~90%), with final scores denoted on a 0−100 scale. Higher scores denote better sleep quality. Continuous data tables were then generated with bi-weekly and minute granularity via linear interpolation. The IS subscale score was then calculated using a weighted average.

Item weightings: To determine coefficients of the weighted average, four clinical sleep experts independently rated the importance of each IS variable against another using a relative weighting (<1 weak, 1 = equal, >1 strong). The process was completed for all binary combinations of IS items (10 total), such as the relative importance of frequency of night wakings compared to duration of night wakings. Each combination of IS items was then consolidated into a system of equations. After solving for each equation, IS coefficients (item weights) were determined.

2.2.2. Parent perception subscale

The PP includes three questions regarding caregiver perceptions of bedtime difficulty, overnight sleep, and overall child sleep problems.

Item scoring: Item scoring in this subscale is based on linear interpretation of responses and mapped to a normative scale from 0 to 100. For example, parent perception of "sleep problems" item is scored from 10 (serious problem) to 30 (moderate problem) to 50 (small problem) to 80 (very small problem) to 100 (no problem). Thus, higher scores denote more positive perception of sleep quality. The total subscale score is then calculated using a weighted average.

Item weightings: To determine coefficients of the weighted average, parent perception of sleep problems is weighted twice that of perception of overnight sleep and bedtime difficulty, based on clinical expert consensus.

2.2.3. Parent behavior subscale

The final subscale PB, includes 11 questions about sleep ecology, including bedtime routine consistency, bedtime, parental behavior at time of sleep onset and following night wakings, and sleep locations at time of sleep onset and following night wakings.
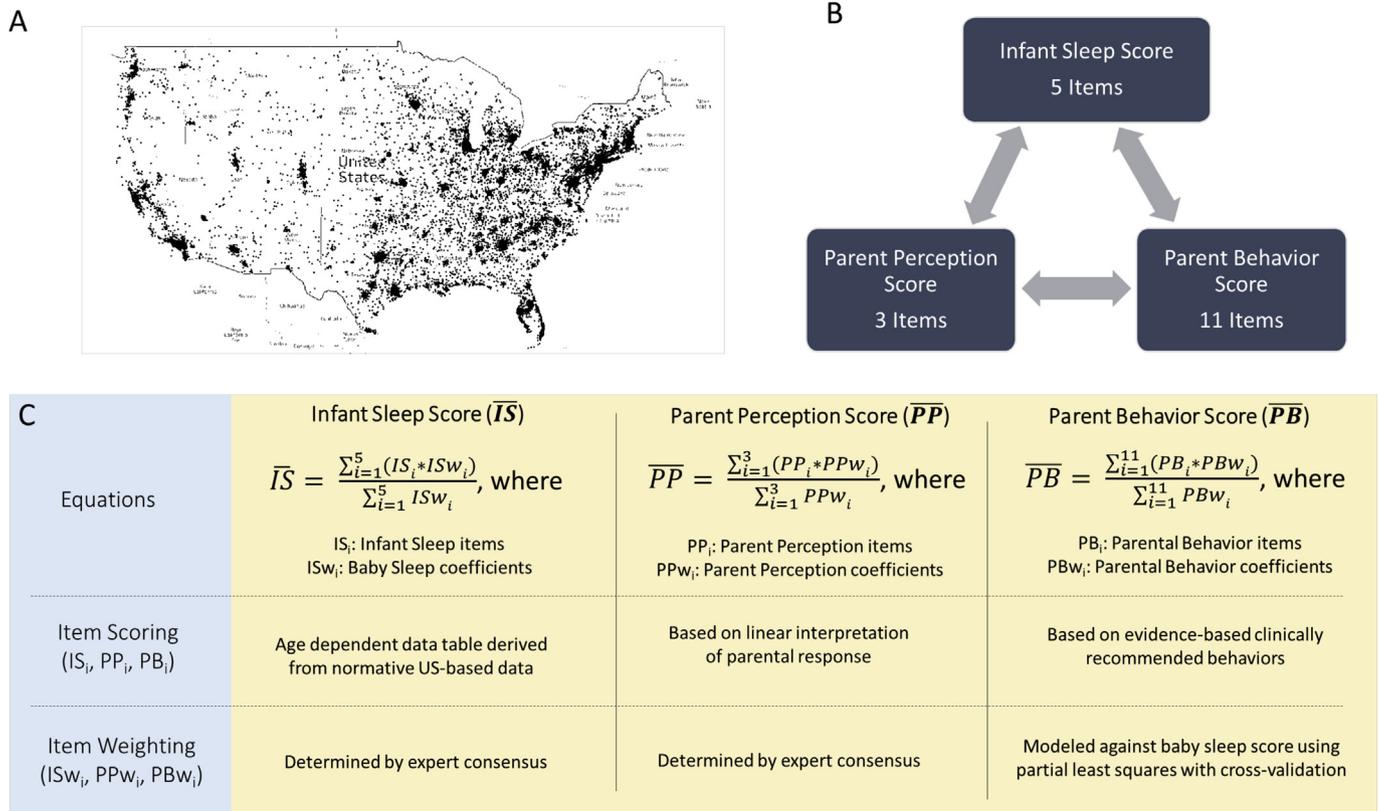
A

B

C

	Infant Sleep Score ($\overline{IS}$)	Parent Perception Score ($\overline{PP}$)	Parent Behavior Score ($\overline{PB}$)
Equations	$\overline{IS} = \dfrac{\sum_{i=1}^{5}(IS_i * ISw_i)}{\sum_{i=1}^{5} ISw_i}$, where IS$_i$: Infant Sleep items ISw$_i$: Baby Sleep coefficients	$\overline{PP} = \dfrac{\sum_{i=1}^{3}(PP_i * PPw_i)}{\sum_{i=1}^{3} PPw_i}$, where PP$_i$: Parent Perception items PPw$_i$: Parent Perception coefficients	$\overline{PB} = \dfrac{\sum_{i=1}^{11}(PB_i * PBw_i)}{\sum_{i=1}^{11} PBw_i}$, where PB$_i$: Parental Behavior items PBw$_i$: Parental Behavior coefficients
Item Scoring (IS$_i$, PP$_i$, PB$_i$)	Age dependent data table derived from normative US-based data	Based on linear interpretation of parental response	Based on evidence-based clinically recommended behaviors
Item Weighting (ISw$_i$, PPw$_i$, PBw$_i$)	Determined by expert consensus	Determined by expert consensus	Modeled against baby sleep score using partial least squares with cross-validation

Fig. 2. The Brief Infant Sleep Questionnaire-Revised (BISQ-R) scoring framework. (A) 33,835 submissions (real-world data sample 1) of the expanded BISQ by caregivers of infants and toddlers (1−36 mos) were analyzed in the US via a publicly-available smartphone application, Johnson's® Bedtime®. (B) Three subscales were created: Infant Sleep (IS, 5 items), Parent Perception (PP, 3 items), and Parent Behavior (PB, 11 items). (C) The scoring algorithm was based on an age norm-referenced system, and each subscale was scored using a weighted average of items.

Item scoring: Scoring of items in this subscale was derived from evidence-based clinically recommended behaviors [1,16]. All item responses were evaluated in an age-dependent manner (eg, multiple feedings throughout the night are age-appropriate for newborns) and mapped to an age-based norm-referenced scale from 0 to 100. Higher scores denote parent behaviors that promote healthy sleep behaviors and independent infant sleep. The total subscale score was then calculated using a weighted average.

Item weightings: To determine coefficients of the weighted average, we utilized the method of partial least squares (PLS). PLS is a multivariate technique for modeling relationships between a set of predictor (X) and response (Y) variables in terms of a set of mutually orthogonal latent factors, or PLS components. For our model, each parent behavior item was denoted as a predictor (X) and IS score was denoted a response (Y). The nonlinear iterative PLS (NIPALS) algorithm was implemented for computing loadings and convergence (JMP Pro software version 13). Acceptance of the coefficient was based on the minimum root mean predicted residual error sum of squares across all ages. A list of all items and associated weighted average coefficients are presented in Supplement Fig. 1.

2.2.4. Total score

A BISQ-R Total Score was also calculated, which is an average of the three subscales.

Scales on each subscale and the total score are scaled from 0 to 100, with higher scores denoting better sleep quality, more positive perception of infant sleep, and parent behaviors that promote healthy sleep. A copy of the BISQ-R and scoring mechanism can be found at www.babysleep.com/BISQ.

2.3. Real world data sample 1

Caregivers of 33,835 infants and toddlers (1−36 months, mean age 7.87 months, SD = 7.28 months; 52.2% boys; 83.0% mothers) from the United States participated in this study. Age distribution was skewed (1−6 months = 19,270; 6−12 months = 7823; 12−18 months = 3171; 18−24 months = 1679; 24−30 months = 1143; 30−36 months = 749). Data were collected from those who completed the BISQ-R on the Johnson's® Bedtime® baby sleep application (app) between 2013 and 2016, a free, publicly available smartphone app for sleep education and management in young children. Depiction of users, based on zip code, indicates representation across the US and is highly similar to population density maps (see Fig. 2A). Data using this participant and data collection methodology have been previously published [10,17], but this is the first presentation of this complete data set. For quality control, replies with inappropriate or extreme data (eg, nighttime sleep duration <5 h or >14 h) were excluded. Similar data cleaning rules were applied for all of the following studies.

2.4. Primary application using real world data sample 2

To validate the scoring algorithm, a second sample of caregivers of 16,531 infants and toddlers (1−36 months; 53.9% boys; 87.9% mothers; mean age 7.31 months, SD = 6.72 months) from the United States participated in this study. Age distribution was skewed (1−6 months = 9966; 6−12 months = 3881; 12−18 months = 1,331, 18−24 months = 656; 24−30 months = 434; 30−36 months = 264). Data were collected from those who completed the BISQ-R on the Johnson's® Bedtime® baby sleep app

between 2016 and 2018. These data have not been previously published.

2.5. Secondary application using web-based data sample 3

Parents of 4595 infants and toddlers (1–36 months; 52.0% boys; 97.1% mothers) completed an online survey in the US and age was equally distributed. All data were collected via BabyCenter.com, a popular parenting website, during May and June of 2006. An online version of the BISQ-R was set as a pop-up screen and invited parents to complete a sleep survey for children aged three years old or younger. Completion of the questionnaire was voluntary, and parents were not offered any compensation or feedback for their participation. Complete information about this sample and results have been previously published [12].

2.6. Tertiary application using longitudinal data sample 4

Assessments were completed at 3, 6, 9, 12, and 18 months across five cohorts of children for a total of 164 mother-child dyads. Families were recruited when their baby was either 3, 6, 9, 12, or 18 months. The BISQ-R was completed by each family every 3–6 months for 4 or 5 months, spanning a time frame of 12–15 months for each child. Thus, mothers completed the BISQ-R on paper at each time point. This study was conducted from 2008 to 2010. Data from a subset of this sample has been published previously regarding the relationship between sleep and social-emotional development [18].

2.7. Tertiary application using randomized control trial data sample 5

Overall, 264 mothers and their young child (6–36 months; 49.6% boys) participated in this randomized control trial (RCT) assessing the efficacy of an internet-based intervention for bedtime problems and night wakings in young children. Complete information about this sample and the results of the RCT have been previously published [19]. Inclusion criteria for the study included that all children must have a parent-identified sleep problem, with all mothers endorsing that their child had a sleep problem that ranged from "small" to "serious," as well as experienced bedtime difficulties. Families were randomly assigned (ensuring equal gender and age) to a control group or one of two intervention groups (online intervention or online intervention/bedtime routine). All families completed a one-week baseline period in which mothers followed their child's usual bedtime practices and then either implemented a prescribed intervention or continued their usual practices. The BISQ-R was completed online at the end of baseline (day eight) and following implementation of intervention (days 15 and 22) regarding the previous week. Note that this research design resulted in parents completing the BISQ-R based on the past week rather than the past two weeks. This study was conducted in 2009, with one-year follow-up completed in 2010.

3. Results

3.1. Algorithm implementation – Real world data sample 1

The scoring algorithm was applied to the real-world data sample 1, collected as the original sample of sleep patterns and behaviors in infants and toddlers. As shown in Fig. 3A, each subscale (BS, PP, PB) and the Total score (T) was norm-referenced based on age (0–100), and cross-comparison between subscales revealed consistent relationships. As expected, there was a moderate relationship between IS and PP subscales ($R^2 = 0.335$, $P < 0.001$) (Fig. 3B). Significantly,

regression coefficients between IS and PB revealed a strong logarithmic increase ($R^2 = 0.899$, $P < 0.001$) (Fig. 3C). This represents that the association between parent behavior and IS is stronger for older infants and toddlers. It should be noted that average scores in this real-world data sample 1 are likely a bit lower than the general population, as 42.5% of parents reported a sleep problem ("do you consider your child's sleep a problem), compared to 20–30% reported in previous studies [1,12]. The complete raw subscale scoring distribution tables are in the supplement (see Sup Fig. 3).

3.2. Primary replication – Real world data sample 2

The scoring algorithm was then applied to the real-world data sample 2, collected as the primary replication sample of sleep patterns and behaviors in infants and toddlers. As shown in Fig. 4A, each subscale and total score (BS, PP, PB, T) was norm-referenced based on age (0–100), and weighted averaged PB coefficients converged with previous partial least squares analysis (Sup Fig. 2). Cross-comparison between subscales revealed consistent and convergent relationships. As expected, there was a moderate relationship between IS and PP subscales ($R^2 = 0.312$, $P < 0.001$) (Fig. 4B). Regression coefficients between IS and PB revealed a strong logarithmic increase ($R^2 = 0.876$, $P < 0.001$) (Fig. 4C). Again, this nicely represents how parent behavior is related to IS in our age dependent scoring algorithm. It should be noted that average scores in this real-world data sample 2 are slightly lower than in real-world data sample 1, a result likely because more parents (52.2%) reported a sleep problem ("do you consider your child's sleep a problem?"). The complete raw subscale scoring distribution tables are in the supplement (see Sup Fig. 3).

3.3. Secondary application – Original extended BISQ web-based ecology study

The scoring algorithm was then applied to the web-based data sample 3, collected as the original extended BISQ web-based ecology study in 2009. As shown in Fig. 5A, each subscale and the total score (BS, PP, PB, T) was norm-referenced based on age (0–100), and cross-comparison between subscales revealed consistent relationships. As expected, there was a moderate relationship between IS and PP subscales ($R^2 = 0.316$, $P < 0.001$) (Fig. 5B). Regression coefficients between IS and PB revealed a strong logarithmic increase ($R^2 = 0.724$, $P < 0.001$) (Fig. 5C). Notably, average scores are higher in this study, a result likely because this sample of sleepers had a more representative prevalence of sleep problems, although this was based on the original BISQ, which utilized a 3-point rather than 5-point scale for the question "do you consider your child's sleep a problem?" The complete raw subscale scoring distribution tables are in the supplement (see Sup Fig. 4).

3.4. Tertiary application – stability of scores over time

The scoring algorithm was then applied to the longitudinal data sample 4. As shown in Fig. 6, each subscale and the total score (BS, PP, PB, T) was norm-referenced based on age (0–100), There is significant individual variability in scores at each time point, with relatively stable scores across the 12- to 15-month follow-up period. The percentile distributions as a function of age for each subscale are in the supplement (see Sup Fig. 5). Note that average scores on each subscale and total score are again higher than in the initial normative study reported above, as this sample ended up having few sleep problems. Thus, only 9.4% of mothers in this study reported that their child had a sleep problem across all time points; a relatively low percentage compared to community-based studies.
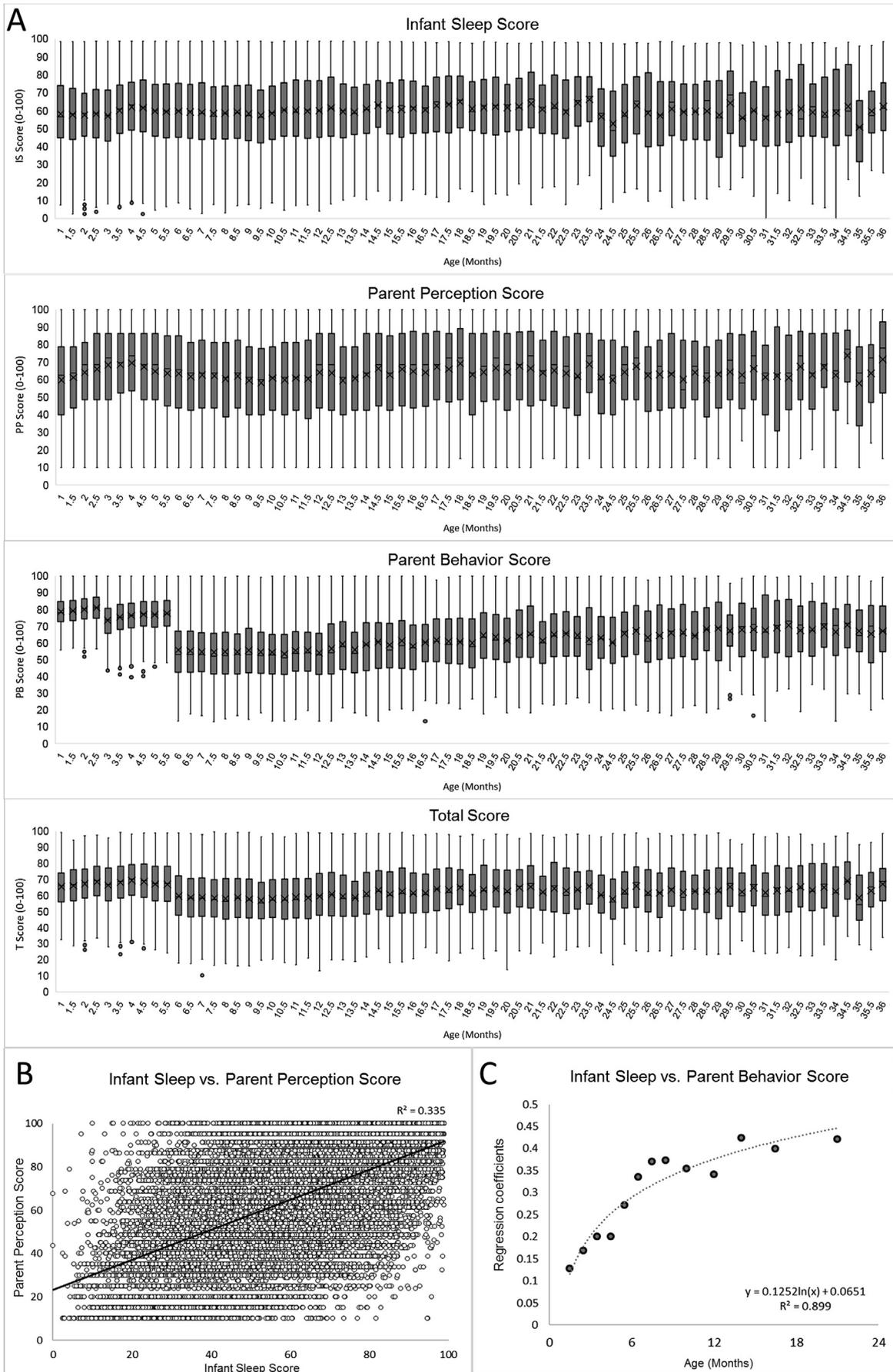
Fig. 3. Scoring algorithm implementation using the real-world data sample 1 (33,835 submissions). (A) Age based norm-referenced scoring for each subscale ranging from 0 to 100 (Infant Sleep-IS, Parent Perception-PP, Parent Behavior-PB, Total-T). (B) Correlation between IS and PP. (C) Regression coefficient correlation between IS and PB as a function of age.
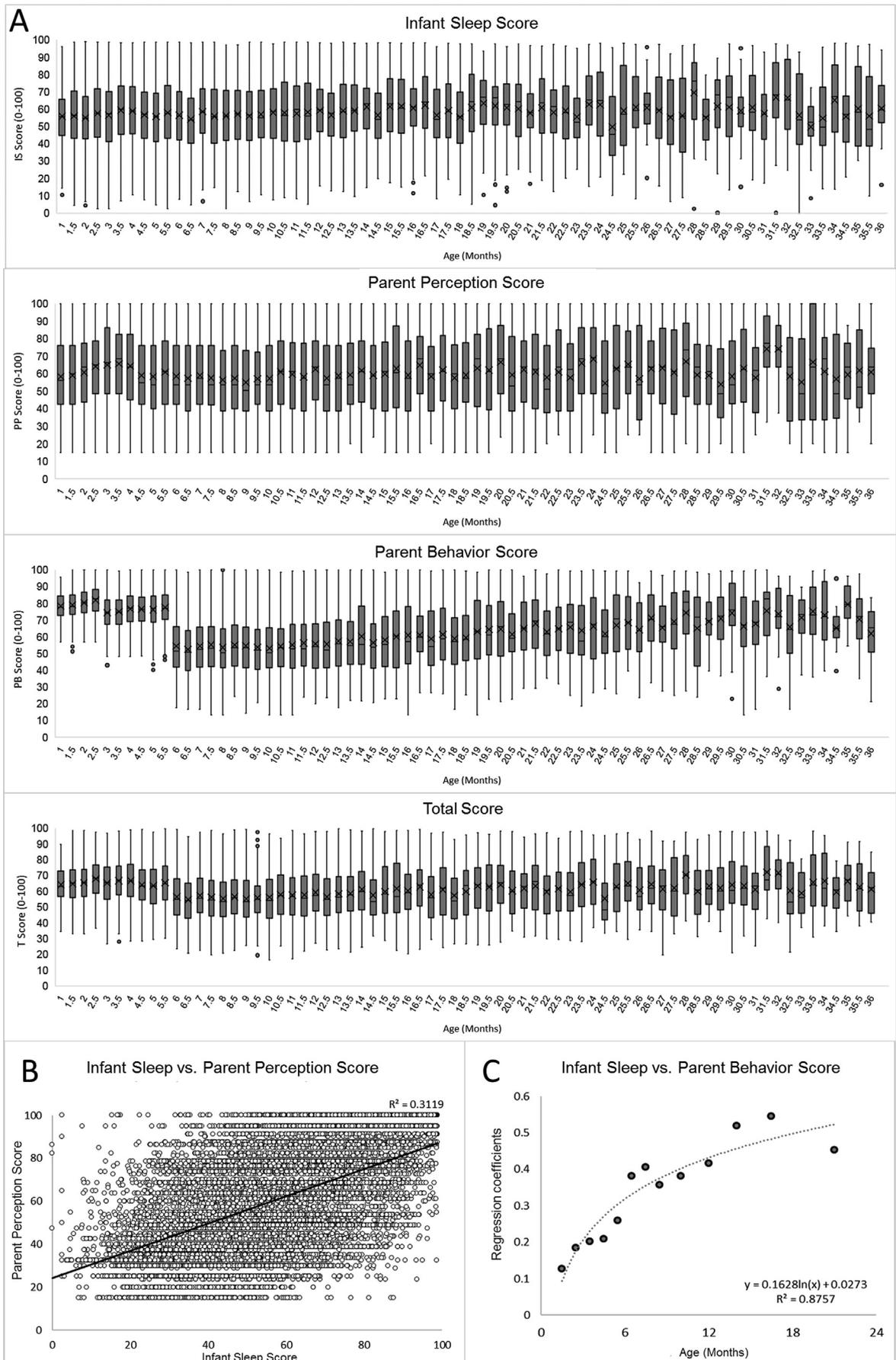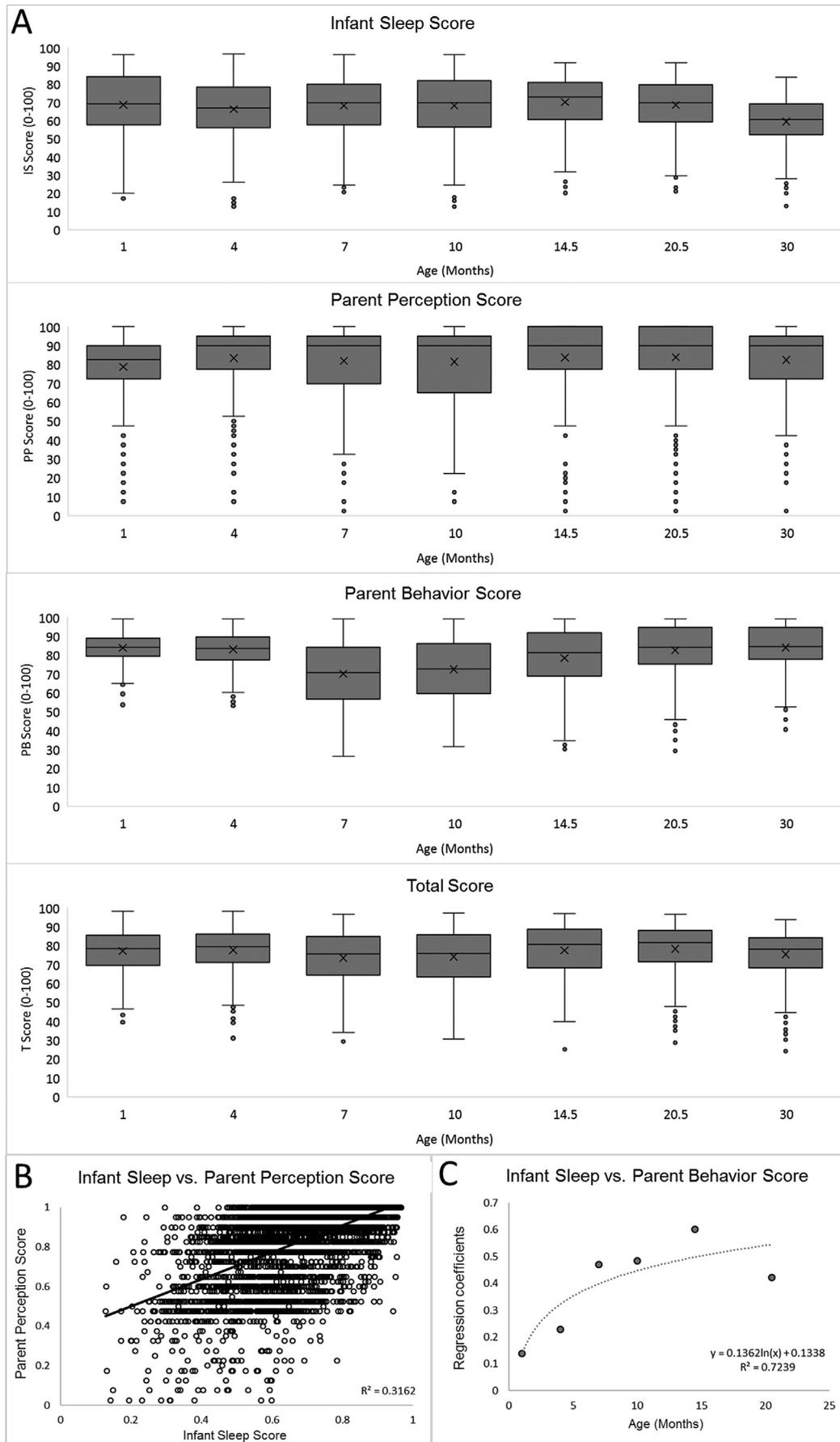
Fig. 4. Primary replication using real-world data validation sample 2 (16,531 submissions). (A) Age based norm-referenced scoring for each subscale ranging from 0 to 100 (Infant Sleep-IS, Parent Perception-PP, Parent Behavior-PB, Total-T). (B) Correlation between IS and PP. (C) Regression coefficient correlation between IS and PB as a function of age.

Fig. 5. Secondary replication using the web-based ecology data sample 3 (4595 submissions). (A) Age based norm-referenced scoring for each subscale ranging from 0 to 100 (Infant Sleep-IS, Parent Perception-PP, Parent Behavior-PB, Total-T). (B) Correlation between IS and PP. (C) Regression coefficient correlation between IS and PB as a function of age.
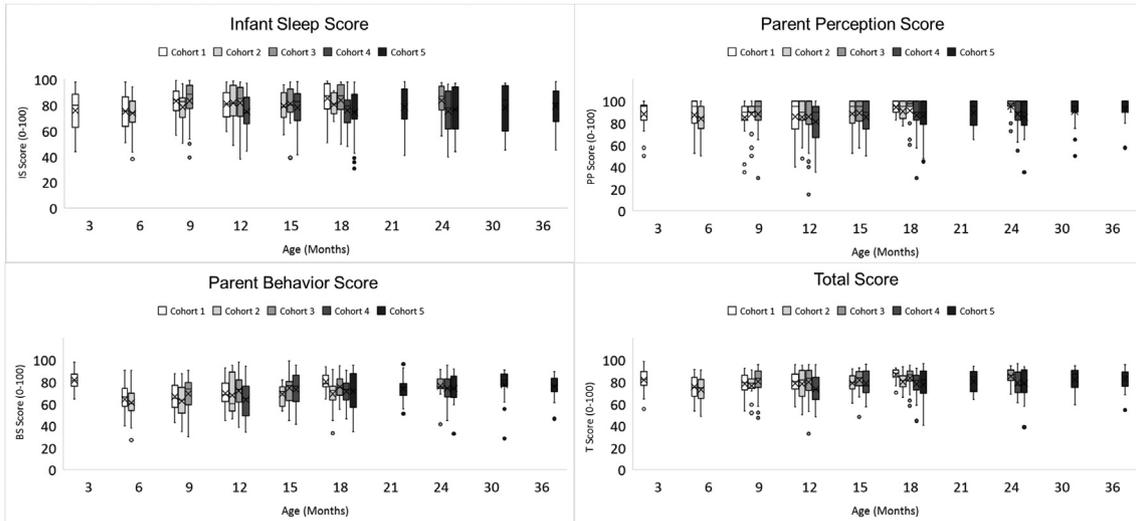
Fig. 6. Tertiary application using the longitudinal data sample 4 (166 mother-child dyads).
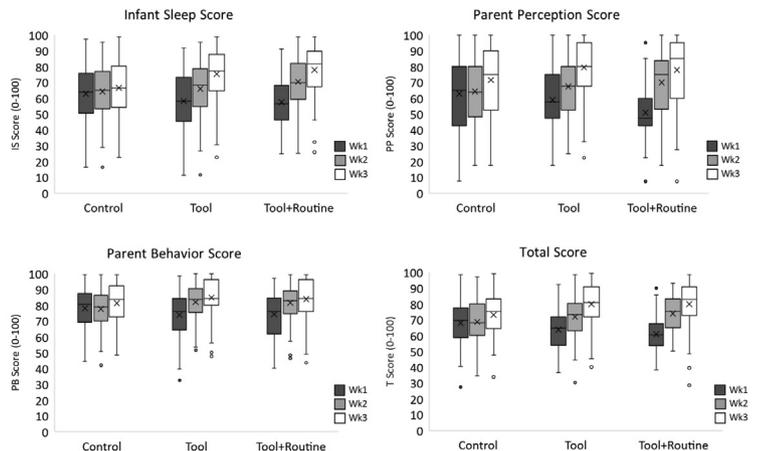

Fig. 7. Tertiary application using the randomized control trial data sample 5 (264 mother-child dyads).

3.5. Tertiary application — Change in response to intervention

The scoring algorithm was then applied to a randomized control trial data (sample 5). In the original study, analyses were conducted on individual items. Data from this study were re-analyzed using BISQ-R scoring for the control and intervention groups. As denoted in Fig. 7, no significant differences across time were found for the subscale and total scores for controls, $p > 0.05$. ANOVA analyses indicated that all subscale scores and the total score significantly improved from baseline to post-intervention for both intervention groups, $p < 0.001$. These results are consistent with those presented in the original paper and reflect the original item-by-item analyses.

4. Discussion

This study aimed at addressing the main limitation of the lack of a scoring system for the BISQ-R, a commonly used measure assessing sleep in infants and toddlers. Three subscales were conceptually created denoting individual aspects of infant sleep — IS, PB, and PP. A norm-referenced age-based scoring system based on these three aspects was thus developed and applied across five different studies, including cross-sectional, longitudinal, and intervention-based studies. Its scoring was found to support

expected relationships between infant sleep and parent behaviors (sleep ecology) and perception. For example, parental behaviors such as parental presence at bedtime and lack of a consistent bedtime routine have been found to be associated with decreased sleep consolidation [3,12] and night wakings associated with parental perceptions of sleep problems [20]. The scoring system also supported stability across time in a longitudinal study, as well as being sensitive to positive outcomes in a randomized clinical trial of the efficacy of an intervention study.

4.1. Limitations and future directions

One of the primary limitations of the newly developed scoring system is that all scoring is based on samples in the United States. Previous studies have shown that there are significant cross-cultural differences in sleep patterns and perceptions of sleep problems cross-culturally [2], utilizing both the BISQ and other assessment measures. Given that the BISQ and BISQ-R have been translated into over 20 languages, efforts are being made to create age-based scoring systems for other countries. As those get developed, they will be made publicly available. Furthermore, there is a potential concern that the development of the scoring was based on a sample that had a higher rate of parent-perceived sleep

problems than the general population. The potential consequence is that calculated scores may be slightly higher than expected, as inclusion of more non-problematic sleepers may have resulted in relatively lower scores for problematic sleepers. This shift, however, does not invalidate the scoring system, as all scores are relative.

An additional limitation is that the item weights, especially for parent behaviors, may be construed as culturally-biased, such as the denotation of higher scores for falling asleep independently. Therefore, those who use this scale in a research or clinical context need to consider individual family and cultural preferences. Furthermore, each subscale score should be considered within the context of the other subscale scores. For example, a relatively low PB score would likely not be considered problematic within the context of a high IS and PP score. Similarly, a focus on subscale scores rather than the total score is recommended in the use of the BISQ-R. For example, studies have found cultural and socioeconomic differences in the overall perception of sleep problems, as well as differences in the relationship between infant sleep outcomes and parental perception of sleep problems [20–22].

Additional future directions are continued validation of the BISQ-R, including construct validity (comparison of clinical and community samples) and convergent/divergent validity, as well as development and validation of clinical scoring guidelines. Note that past studies of the BISQ and BISQ-R [11,13] have supported its reliability, including high test-retest reliability, interobserver reliability, and intraobserver reliability. Validation of this measure has also supported its ability to distinguish between clinical and control samples, document expected developmental changes, and, demonstrate high construct validity (associations with parental diary and actigraphy).

5. Conclusions

Overall, the development of a norm-referenced age-based scoring system of the BISQ-R enables clinicians and researchers alike to assess infant and toddler sleep. The availability of a scoring system for this already highly used measure increases the utility of using it in both clinical and research settings. A publicly available copy of the BISQ-R and online age-based scoring can be found at www.babysleep.com/BISQ.

Financial disclosure statement

Funding source

Acknowledgments

Conflict of interest

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.sleep.2019.05.010.

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